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Integrating Qualitative Techniques in Model Development: a case study using the framework approach

Running title: Application of framework approach in model development

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ABSTRACT

Background: Despite their potential, there is limited uptake of formal qualitative methods in model development by modellers and health economists. The aim of this case study is to highlight in a real-world context how a qualitative approach has been applied to gain insight into current practice (delineating existing care pathways) for typhoid fever in Ghana which can then assist in model structure conceptualisation in a model-based cost-effectiveness analysis.

Methods: The perspectives of a range of healthcare professionals working in different settings and across different practices in the Eastern region of Ghana were captured with a self-administered survey using open-ended questions and analysed using the framework method.

Results: A total of 51 completed questionnaires were retrieved representing a 73% response rate. It was found that two main care pathways for typhoid fever exist in Ghana and there was no consensus on how a new test might be applied to the existing pathways.

Conclusion: The two settings in Ghana have different care pathways and any cost-effectiveness analysis should consider the alternative pathways separately. This study has demonstrated that framework analysis is a qualitative methodology that is likely to be accessible and feasible across a wide range of health economic settings.

Key points for Decision Makers

- Delineating care pathways during model development can be difficult and qualitative methods play a potentially key role in enhancing this process.
- Integrating qualitative methods in model development can potentially enhance the validity as well as generalizability and credibility of models.
- Framework analysis is a simple but systematic qualitative approach that can potentially enhance the process of model development.

Key words: qualitative techniques; framework method; model development; cost-effectiveness; care pathways; typhoid fever

1.0 INTRODUCTION

There exist challenges for health economic evaluation of medical tests [1]. Direct research required for their evaluation is lacking [2]. Therefore, test evaluation in most cases generally involves modelling of test-treat pathways by integrating evidence from studies evaluating one or more aspects (test accuracy, test performance, treatment effectiveness) within the care pathway [3]. An informative economic model will require knowledge of care pathways if it is to represent appropriately the disease, persons not having the disease, misclassifications, and be fit for the purpose of decision making [4]. Therefore, delineating the care pathway and any existing variations may be a key information requirement to effective test evaluation [5].

Delineating the test-treat pathway can be difficult and qualitative methods can play a potentially key role in enhancing this process [6-10]. For example, they can be used to facilitate and capture the perspectives of all of the different types of experts knowledgeable on the topic of interest [11], including capturing variability in practice when defining care pathways to assist in model structure conceptualisation [12]. Variability may arise from clinicians and patients involved at different points in a care pathway and in different settings/locations. Capturing variability can potentially assist in improving the validity as well as generalizability and credibility of models. There are examples of different qualitative research methods used for model development including the Delphi technique [12,13], focus groups [8], and stakeholder workshops [14]. Despite their potential, there is limited uptake of formal qualitative methods in model development by modellers and health economists [15]. One explanation for their limited uptake is lack of familiarity of their potential to assist with model development and a general assumption that qualitative methods are resource intensive.

The diagnosis of typhoid fever represents an area of unmet need in Ghana [16]. The investigative tool most often used is the Widal test (a serological test). Typhoid fever has been over diagnosed based on the Widal test [17], and the WHO recommended investigative

tool (blood culture) [18] has its limitations. Low sensitivity (the ability of the test to correctly classify the proportion of people with disease who will have a positive test result) remains a limitation to this diagnostic method (as low as 40%) [19]. The potential impact is that, the disease will be missed in a lot of patients (i.e., a high false negative rate) resulting in lost opportunity for treatment, which might lead to increased morbidity and mortality. Furthermore, culture testing requires resources (trained personnel and equipment) which may not be available in primary health care facilities in Ghana (as in other resource-constrained settings) and results take 2-3 days. This may result in diagnosis being delayed or overlooked, and patients may end up with complications if treatment is postponed or patients are lost to follow up [20]. Typhoid fever clinically resembles other febrile conditions such as malaria and is easily misdiagnosed without laboratory confirmation [21], which could lead to an overprescribing of antimalarial therapies associated with a huge economic impact on the Ghanaian economy. Consequently, concerted efforts are being made to develop a clinically effective and cost-effective rapid diagnostic test that will aid fast accurate diagnosis and management of typhoid fever cases in Ghana.

Identifying care pathways for typhoid fever in Ghana, their variations, diagnostic challenges associated with these (which are likely to result from the different levels of care) and the potential placement and role (replacement, triage or add-on) of a new test on existing care pathways are key to the effective evaluation of the true value of a new test. This requires novel research because records cannot be relied on to obtain such relevant information. The characteristics of a test: accuracy (sensitivity, specificity) and processing requirements (for example expertise, equipment needed, and time taken) will determine the best placement and role on a testing pathway. A test intended to be a replacement test is expected to be more accurate, less invasive for the patient and yields quicker results. A test intended to be a triage test may be less accurate than existing ones, should be non-invasive, simple to perform and

cheap. A test may be used as an add-on test at the end of a testing pathway because although it is more accurate, it is also more invasive or expensive. Consequently, its application is reserved for specific patient subgroups in which the test is deemed a final resort [22].

The aim of this case study is to highlight in a real-world context how a qualitative approach has been applied to gain insight into current practice (delineating existing care pathways) for typhoid fever in Ghana and the potential role of a hypothetical test on the pathway, which can then assist in model structure conceptualisation in a model-based cost-effectiveness analysis. Specifically, the aim is to describe the existing care pathway for typhoid fever in Ghana and any variations, explore reasons for variations in existing care pathways for typhoid fever in Ghana, and to capture the views of a range of healthcare professionals working in Ghana on the potential role of a hypothetical new rapid test for typhoid in the care pathway they work in.

2.0 METHODS

The perspectives of a range of healthcare professionals working in different settings and across different practices in the Eastern region of Ghana were explored to answer the questions: what is the test-treat pathway for typhoid fever in Ghana? Are there variations in the existing test-treat pathway? And what is the potential role(s) of a new test on the existing pathway in Ghana? One region was chosen as a case study because the literature on the structure of the healthcare delivery system in Ghana indicated that variations in practice were likely to result from the different levels of care (which is the same across all the regions) rather than resulting from the individual regions. The Eastern region was selected because the Principal Investigator (PI) worked and lived in this region and is familiar with the region thus it was comparatively less resource intensive to obtain the data required because of proximity. A purposive, maximum variation sampling approach (which involves approaching all those who are knowledgeable on the topic on interest and can give potentially different

perspectives) was undertaken [11]. The results were compared between settings as this was considered the main factor likely to drive any variations in the findings.

2.1 Participants

To be eligible for inclusion in the study, the participant had to be a clinician or medical assistant who worked in a healthcare provision facility (Health Centre, Polyclinic, hospitals, etc.) in the Eastern region of Ghana. Clinicians and medical assistants were chosen to participate in this study because they were knowledgeable in the topic of interest and thus were in a better position to provide valuable information to aid this study. Participants of all ages and genders were included to avoid sampling bias.

2.2 Data collection (March – July 2017)

Some level of pragmatism was required to choose the most appropriate approach to collect data. In-depth interviews were not possible because of participants' time constraints. Therefore a self-administered survey using open-ended questions was used [23]. Acknowledging that responses were likely to lack depth, and a survey would not allow probing by the researcher or checking of the participants' understanding of the questions, a well written questionnaire was required to elicit adequate valuable information. A questionnaire was piloted in a different region (Greater Accra) and revised based on responses into the final version used in this study. With ethical approval from the University of Birmingham (ERN_16-0947) and from the Ghana Health Service Ethical Review Committee (GHS-ERC: 03/12/16), medical superintendents of various health facilities were contacted to arrange a meeting between the researcher and the clinicians and/or medical assistants working at their facilities. During these meetings, the aims and the expected outcomes of the study were explicitly explained to them and interested persons were recruited into the study after having gone through the informed consent process and signed the consent

form. Groups of recruited participants in each institution were given the opportunity to go through the questionnaire to ensure they understood the questions, and explanations were offered to clarify issues where necessary. Questionnaires were coded for easy identification of the facility and its location within the region to enable easy analysis of the data. Participants were anonymized. A total of 70 self-administered questionnaires were distributed to participants by the researcher in person.

The following themes were generated *a priori* by the research team and informed the content of the questionnaire: presumptive treatment views (treatment of clinically suspected cases without, or prior to, results for confirmatory laboratory test), diagnostic test(s) used, test negative management (care plan for those with negative test result), test positive management (care plan for those with positive test results), prescribed antimicrobial for positives, dosage of antimicrobial, assessment of test-treat outcome and intended role of new test. All the themes except for "intended role of new test" were generated to gain insight into current practice by considering them collectively and thus are reported and discussed under the section "delineating existing test-treat pathway and reasons for variation". "Intended role of new test" was examined to capture views on the potential role of a hypothetical new test, and where it should be placed on the testing pathway to improve current practice. The chosen role of a new test implies the necessary properties it should possess. Participants were asked to choose a statement which best describes the role of a hypothetical new rapid diagnostic test for typhoid: (a) to test patients first to decide on who should receive further testing with the test you use now (triage); (b) as the main investigative tool without further testing with the test you use now (replacement); or (c) to provide additional diagnostic information after testing with the test you use now (add-on). The questionnaire is shown in Appendix 1.

2.3 Data analysis

Framework analysis was adopted as the analytical framework for analysing the data collected in this study [24]. This method of analysis belongs to a wider group of analysis methods (termed thematic analysis or quantitative content analysis) which seeks to identify, analyse and report patterns within data [25]. The data collected is based on pre-defined themes (deductive) rather than inductive (emerging from the data). The distinctive attribute of this method is the matrix output, which provides a systematic structure into which data can be charted (inputted) to enable in-depth analysis [26]. It consists of rows, columns and cells of summarised data. In this case study, rows referred to participants, columns referred to pre-defined themes, and cells of summarised data referred to responses from participants. Framework analysis was chosen due to its deductive approach that makes it especially suited to research that has specific data needs set in advance [27]. In this case study, it allowed for pre-defined themes to be developed which shaped the nature of the data collection. In addition, framework analysis was selected because it is intuitive and allows for easy recognition of patterns in the data set once a matrix has been developed [24]. Also, the systematic approach embedded in the framework method of analysis ensures that the rigour of the analytical process is maintained, thereby enhancing the credibility of findings [28]. Figure 1 illustrates the procedure for analysis using the framework approach.

Written responses retrieved from participants were first converted into electronic form by the principal researcher and read repeatedly by members of the research team to ensure familiarisation with the data. This provided a good opportunity to get immersed in the data set and have a better recollection of key information [25]. The textual data was colour coded (by two members of the research team individually) to classify all of the data set according to the pre-defined themes. There were no emerging new themes from the survey, thus, the pre-defined themes generated were then developed into a working analytic framework; a matrix

consisting of rows and columns into which data was charted to enable in-depth analysis [26]. The “analytical framework matrix” is shown in Appendix 2.

3.0 FINDINGS

3.1 Demographics of participants

A total of 51 questionnaires were retrieved representing a 73% response rate. Non-respondents were all participants working in primary hospitals and time constraint was the main reason given for non-response. Twenty responses were from participants working in secondary care and thirty-one from participants in primary care. The name, gender, and age of the participants were not captured to ensure participant anonymity.

3.2 Delineating existing test-treat pathway and reasons for variations

In terms of gaining insight into current practice two main care pathways were delineated from the analysis of the matrix; the “culture care pathway” (suspected typhoid cases have culture i.e., blood, stool or urine) and the “Widal care pathway” (suspected typhoid cases have Widal test) (Fig 2&3). Underpinning this variation was the type of healthcare provision facility in which the participants were recruited from. It was noted that the “Widal care pathway” was predominantly followed in primary hospital facilities (typically district level facilities that provide the most basic care) whereas the “culture pathway” was predominantly followed in the secondary hospital facility (typically regional hospitals which are an upgrade of primary hospitals in terms of infrastructure and resources). However, there were two exceptions to this pattern. Blood or stool culture was stated by **participant 50** (P) as the diagnostic tool used even though the participant was recruited from a primary hospital facility and **participant 31**(S) stated the use of the Widal test even though the participant was recruited from a secondary hospital facility (letters P and S in parenthesis indicate whether a

participant was recruited from primary care or secondary care respectively). **Participant 31**(S) stated, *"it is the commonest and simple to do"* as the reason for using the Widal test. This quote is suggestive of the fact that the Widal test could be available in secondary hospitals even though culture might be the preferred test in such settings. **Participant 50**(P) **stated**, "*it is readily available in our lab"* as the reason for employing culture.

The management aspect of the two care pathways following testing with either the Widal test or culture was found to be similar. In both pathways, test negative patients were investigated further for other conditions especially malaria and treated according to the final diagnosis. For example, **participant 16**(P) stated, *"when negative, we check for other conditions like malaria and treat them"*. **Participant 19**(P) stated, *"I look out for other conditions presenting like typhoid fever in typhoid negative clients and treat"*. **Participant 35**(S) stated, *"when negative I investigate underlying cause of symptoms and treat appropriately"*. And **participant 38**(S) stated, *"when negative, I investigate further to get the specific condition"*. Typhoid positive patients were noted from the analysis to be treated with antibiotics mainly oral ciprofloxacin at a dose of 500mg every twelve hours for a period of seven to fourteen days. After this, they were reviewed and in most cases re-tested to ascertain whether the test-treat process was successful in improving the patient's health. For example, **participant 19**(P) stated, *"I treat typhoid positive patients with oral ciprofloxacin. Dosage: Tb Ciprofloxacin 500 mg bd* 7-14 days"*. **Participant 25**(P) stated, *"positives: I treat with ciprofloxacin. Dosage: Tb Ciprofloxacin 500mg bd *14 days"*. And **participant 32**(S) stated, *"I give oral antibiotics medication for positive patients. Dosage: Tb Ciprofloxacin 500 mg bd* 10 days"*. It was noted that, the choice of oral ciprofloxacin was because it is the first line drug to manage typhoid fever and for other reasons such as its efficacy against the causative organism, availability and cost. For example, **participants 31**(S) **and 51**(P) stated, *"it is the first line drug to manage typhoid"*. And **participant 33**(S) stated, "*because of its efficacy and*

sensitivity against the causative organism Salmonella typhi". However, it was noted that some participants prescribed alternative antibiotics to oral ciprofloxacin. For example **participant 1**(P) stated, *"positive cases are put on treatment. Dosage: Tb Metronidazole 200-400mg tds* 7days"*. **Participant 4**(P) stated, "*treat positives with antimicrobials. Dosage: Tb Cefixime 200mg bd*7 days"*. **Participant 8**(P) stated, *"I give antibiotics for test positive cases. Dosage: Tb Azithromycin 1g daily* 7days"*. And **participant 30**(S) stated, *"positives: I give antibiotics. Dosage: Tb Cefixime 400mg bd*10-14 days"*.

Of further interest was the views of participants on applying presumptive treatment for typhoid fever. The majority of participants were averse to presumptive treatment, because they expressed the opinion that this may lead to treatment failure and the development of antimicrobial resistance. For example, **participant 18**(P) stated, *"it could lead to treatment failure and antimicrobial resistance"*. **Participant 28**(S) stated, *"it can cause antibiotic resistance"*. **Participant 11**(P) stated, *"presumptive treatment may be prophylactic but can cause antimicrobial resistance"*. And **participant 1**(P) stated, *"typhoid fever presents like malaria and presumptive treatment may lead to treatment failure"*. A very few advocated for presumptive treatment, but then only under certain circumstances. For example, **participant 50**(P) stated, *"since culture usually takes 72 hrs, presumptive treatment can be started"*. **Participant 30**(S) stated, *"this is a good intervention to prevent complications since the most precise blood/stool cultures are not available in most district and health centres"*. And **participant 23**(P) stated, *"Widal test is not solely reliable hence treatment can be made if test is not done but clinical features are present"*. From these quotes, it can be concluded that presumptive treatment appears to be used because of delays in diagnosis or concerns about the accuracy of the test or lack of availability.

3.3 Perceived role of new test

Twenty-eight participants from different levels of care stated that they saw themselves using a new test, *"to provide additional diagnostic information after testing with the test they were using currently (add-on)"*. Sixteen participants from the different levels of care stated that they saw themselves using a new test, *"To test patients first to decide on who should receive further testing with the test you use now (triage)"*. And six participants from the different levels of care stated that they saw themselves using a new test, *"as the main investigative tool without further testing with the test they were using currently (replacement)"*. This observation was further explored according to health care setting and it was found that there was a similar trend in the response by health care setting.

The chosen role of a new test implies the necessary properties it should possess. Thus, participants' views on what test properties the new test should possess were explored. The majority of the participants wanted the new test to be more sensitive followed by those who wanted it to be more specific (the ability of a test to correctly classify the proportion of people without the disease who will have a negative test result) with **participants 8**(P) **and 30**(S) stating that they wanted the new test to be more sensitive and more specific compared to the test they were currently using. Some other properties as stated by the participants were improved reliability, low cost, greater availability, better accuracy, the ability to use other fluids apart from blood, easy to use, fast results, and high predictive values. It was noted that for some participants, their views on what test properties a new test should have did not correspond with their choice of the role of the test. For example **participant 37**(S) stated *"better accuracy, low cost, accessibility and ease to perform"* as the properties of the new test but wanted the test to be used as an add-on to culture. However, it is intuitive from the stated properties that this test should ideally be placed before culture in the care pathway to

expedite clinical decision making and enhance prompt implementation of an appropriate effective treatment.

4.0 DISCUSSION

An in-depth understanding of test-treat pathways is key to fully capturing the potential health economic impact of medical tests on patient relevant outcomes, and qualitative methods play a potentially key role in enhancing this process despite their limited uptake in practice. This case study focussed on how a qualitative approach has been applied to gain insight into current practice for typhoid fever in Ghana and the potential role of a new test on the care pathway in order to improve current practice. It was noted that two main care pathways for typhoid fever in Ghana exist (Widal and culture pathways) and the majority of the participants in this study were averse to applying presumptive treatment to typhoid fever. Also, it was noted that the majority of participants saw themselves using a new rapid test if it was introduced into clinical practice as an add-on test followed by those who anticipated using it for the purpose of triage, with very few of the participants using it as a replacement to the test they were currently using.

The identification of two main pathways in this study was primarily due to variations in laboratory diagnostic capacity of healthcare facilities from which the participants were recruited. The Widal test is a simpler technology to employ compared to culture which requires resources (trained personnel and equipment) and therefore was more likely to be employed in primary hospitals which lack resources whereas culture was more likely to be employed in secondary hospitals. Although the majority of participants were averse to applying presumptive treatment to typhoid fever, a few advocated for presumptive treatment and for valid reasons such as delays in diagnosis or concerns about accuracy of test or lack of availability. These circumstances reflect the characteristics of current practice and therefore a

new test would need to be "better" in these respects for healthcare professionals in Ghana to use if current practice is to be improved. The test should have a quick turnaround time, at least as accurate (as both tests) if not more accurate and should be accessible. Improved reliability, low cost, greater availability, better accuracy, the ability to use other fluids apart from blood, easy to use, fast results, and high predictive values were mentioned as the properties that a new test should have to improve current practice. These findings are key information that need to be taken into consideration in any effort to develop a new test to be used in the Ghanaian setting. The use of alternative antimicrobials to ciprofloxacin as noted from this study demonstrates the potential for waste in current practice, indicating the need for standardisation in, or adherence to, the treatment regimen for typhoid fever patients.

Furthermore it was noted that regardless of the setting, majority of participants saw themselves using a new rapid test if it was introduced into clinical practice as an add-on test, followed by those who anticipated using it for the purpose of triage, with very few of the participants using it as a replacement to the test they were currently using. However, one would have expected the responses to vary depending on the care pathway in which the participant worked. Underpinning this viewpoint is the fact that, if the main limitation of the Widal test as acknowledged by the participants who follow the "Widal care pathway" is unreliability of the test, then a majority of them should be more inclined towards using a new test which is intended to be more reliable as their main investigative tool rather than as an add-on to the test. The argument might be that the very limitation of the Widal test is the reason why the new test should be used to provide additional diagnostic information. However, if the new test is accurate enough to be used to confirm diagnosis after the Widal test, then it is intuitive that it will replace the Widal test to save money (although it is acknowledged that a cost-effectiveness analysis comparing both tests will be needed to make such a decision). Also, for those participants who follow the "culture pathway" one would

have expected the majority of them to be more inclined towards using the new test for triage purposes. The reasoning is that, if culture takes 2-3 days (causing potential treatment delays), then it is intuitive that patients would be tested first with the proposed test and test positive patients treated promptly whilst test negative patients could be further tested by having their samples cultured. However, it was not possible to explore the reasoning behind the participants' choices (which could be lack of understanding, perception of the accuracy of the existing tests, scepticism about the reality of a new test, acceptability and accessibility of new test).

The findings from this survey can inform the subsequent modelling process in a number of ways. First, delineated test-treat pathways can inform and assist in defining the boundaries and structures of the individual models needed to effectively evaluate the true value of a hypothetical test. It will inform the model structure to appropriately represent clinical practice. These findings indicate that the settings in Ghana are different depending on the level of care, and the implication is that any cost-effectiveness analysis should consider these alternative pathways separately. Second, the findings of the study have assisted in identifying the appropriate comparator test strategies required for the cost-effectiveness study. The potential cost-effectiveness of the hypothetical test should be evaluated by comparing it with each comparator testing strategy separately. This will inform decision making on how the new test should be used in each care pathway, i.e., should it replace the Widal test or should it be used as a replacement, triage or add-on test to blood culture? Furthermore, the intended role of a new test in a care pathway should be clearly defined in the model to enable the assessment of the test in that role. The study findings indicate there was no consensus among the participants on what this role should be, suggesting the need to evaluate the test in all the possible roles in each pathway. It is not clear whether these responses were due to lack of understanding. However, this does not undermine the need to evaluate all possible roles of a

new test. Figures 4, 5 & 6 shows the various model structures evaluating all possible roles of a new test.

Adopting the framework method has worked well for this particular case study. The relatively well structured and deductive approach to questioning enhanced the efficiency of the data collection process because it allowed for the information requirements to be well specified in advance which shaped the nature of the data to be collected. Furthermore, the matrix output allowed patterns to be more easily identified. For example, it was easy to identify the test-treat pathways once the matrix had been developed and data charted into it. An open-ended approach to questioning also allowed the identification of unanticipated issues. An example is the observation with regards to the use of alternative antibiotics to ciprofloxacin. Also, adopting the framework approach allowed for the perspectives of fifty-one participants working in different settings/locations and across different practices (rather than a few informants) to be captured and analysed to gain in-depth understanding of current practice. The implication is that, it could potentially enhance the validity, generalizability and credibility of findings of any modelling study based on the delineated pathways.

Furthermore, many techniques that might be used to elicit test-treat pathways may not be feasible due to time and resource (money and skill) constraints. For example, there are challenges such as the practicality of organising and running face-to-face discussions in focus groups or gaining the inputs of multiple clinicians with busy schedules by asking them to complete questionnaires in a series of rounds as in the Delphi approach. However, as has been shown in this study, framework analysis is an example of a qualitative methodology that is likely to be accessible and feasible across a wide range of health economic settings. The framework method of analysis can in principle be adapted for the analysis of different forms of textual data (e.g., interview transcripts, responses to a questionnaire, meeting minutes and field notes from observations) to produce highly structured outputs of summarised data across

a range of research areas [29]. The key aspect of framework analysis within qualitative research is that it is deductive and allows the researcher to collate qualitative information within pre-defined categories or themes. This is one of the key characteristics of framework analysis that potentially makes it more accessible to quantitative researchers. Thus for this method to be beneficial in any specific health economics context, it is necessary for the researcher to be clear from the outset the issues to be explored (such as the purpose of the model, the type of model to be developed, what elements are heterogeneous etc.). This will inform the most appropriate questions to ask during data collection in order to collect relevant data. Once data has been collected and converted into textual form, the framework method can be applied to analyse the data set to generate important outputs that will appropriately inform the model development process. It is worth mentioning that when reporting qualitative research finding, usually the researcher describes and interprets quotes as they are not self-explanatory without context. However as seen in this study, a survey rather than interviews was conducted and therefore there is limited interpretation. This is due to the nature of the questions asked, eliciting responses that were quite straight forward and requiring less interpretation.

The literature suggests two main ways in which qualitative methods can be applied to the model process. Indeed, there are several studies where the benefits of the application of qualitative methods in model development have been highlighted. For example, Kaltenhaler et al [7] reviewed responses of the evidence review groups (ERGs) (of the National Institute for Health and Care Excellence (NICE)) to manufacturers' model submissions using documentary analysis. They identified several recurring concerns raised by ERGs and made some recommendations to assist manufactures to improve the quality of their submissions. In this instance, a qualitative approach was used to identify and understand areas of concern that required further guidance to inform general and future modelling processes. The Delphi

technique has been demonstrated by Sullivan and Payne [13] and Iglesias et al [12] as a means to gain consensus and collate experts' opinions on certain aspects of a model structure, while the focus group approach for the conceptualisation of models structure has been demonstrated by Roberts et al [8], and Squires et al [14] thus demonstrating the use of stakeholder workshops to conceptualise model structure. Furthermore, there is an increasing reliance on expert judgement as a useful source of data for economic models [13], particularly in early-modelling studies and in cost-effectiveness studies of technologies conducted in the absence of data from randomised controlled trials. Expert judgement is relied on not only to structure and populate a model but also on other important parameters (such as test accuracy) which have an impact on care pathways. Qualitative approaches provide a robust and explicit way to properly elicit their opinions. Clearly, the benefits of using qualitative approaches to inform model development cannot be relegated to the background.

The limitations of this study include the fact that only clinicians and medical assistants were sampled. Other relevant stakeholders such as patients [30] and policy makers could have also been sampled because they may have held different views or added further considerations. Furthermore, because the interviews were not personally conducted, it was not clear whether the participants actually understood all the questions and it was not possible to probe their responses. This was a particular deficiency in this study when capturing valid participants' views about the anticipated role of a new test.

5.0 CONCLUSION

Delineating care pathways during model development can be difficult and qualitative methods can play a potentially key role in enhancing this process. Integrating qualitative methods in model development can potentially assist in improving the validity as well as generalizability and credibility of models. As shown in this case study, qualitative methods

lend themselves well to facilitating the involvement of the different types of experts knowledgeable on a topic on interest and working in different settings (rather than a few informants) to be captured and analysed, thereby potentially enhancing the credibility of the models developed based on the findings.

The qualitative method applied in this study is a simple but systematic approach that can potentially enhance the process of model development. As shown in this study, it provides an alternative way to eliciting responses from relevant stakeholders without having to run face-to-face discussions or by asking them to complete questionnaires in a series of rounds which could be very tasking and time consuming. Thus, by adopting this method, modellers will avoid such additional burden associated with other methods (a possible explanation for the reduced uptake of formal qualitative approaches in model development) whilst still being able to collect and analyse such in-depth information to gain insight into current practice.

One of the defining features of the framework method as highlighted in this study is that, it can be adapted for the analysis of different forms of textual data and it provides explicit steps to follow to produce highly structured outputs of summarised data. Once the matrix has been developed and data charted into it, recognition of patterns in the data set becomes quite straightforward because it becomes easier to compare data across cases and within cases. Therefore, this useful approach could be adopted by health economists and modellers who may want to undertake such studies because of their potentially important role in enhancing model credibility.

Clearly, qualitative methods can potentially assist in improving the model development process and we suggest that those working in this field consider the opportunities that qualitative methods provide. Qualitative investigation is a step beyond the usual informal discussions with clinicians that informs modelling.

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Data Availability Statement

The datasets generated during and/or analysed during the current study are available from the corresponding author on reasonable request.

Author Contribution

All authors take full responsibility for the integrity of the data and the accuracy of the data analysis. SF, CD, AJ and PB conceived and designed the study. The data were acquired by SF and JN but analysed by all authors. All authors participated in the development of this manuscript and gave final approval before submission. **Compliance with Ethical Standards**

The authors Samuel N Frempong, Clare Davenport, Andrew John Sutton, Justice Nonvignon and Pelham Barton, did not receive any financial support and have no conflicts of interest.

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